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Original Article

Sleep disturbances in preschool age children with cerebral palsy: a questionnaire study

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ABSTRACT

Objectives: The study aimed to analyze (i) the prevalence of sleep disorders in pre-school children with cerebral palsy (CP) using the Sleep Disturbance Scale for Children (SDSC), (ii) the possible association with motor, cognitive and behavioral problems, and (iii) the possible differences with typically developing children matched for age and gender.
Methods: One-hundred children with CP (age range: 3–5 years, mean: 3.8 years) were assessed using the SDSC, the Gross Motor Function Classification System (GMFCS), the Wechsler Preschool and Primary Scale of Intelligence, and the Child Behaviour Check List (CBCL) to assess sleep, motor, cognitive, and behavioral problems, respectively. Further 100 healthy children matched for age and sex were assessed using the SDSC.
Results: An abnormal total sleep score was found in 13% of children with CP while 35% had an abnormal score on at least one SDSC factor. SDSC total score was significantly associated with pathological internalizing scores on CBCL and active epilepsy on multivariate analysis. CP group reported higher significant median scores on SDSC total, parasomnias, and difficulty in initiating and maintaining sleep factors.
Conclusions: In pre-school children sleep disorders are more common in children with CP than in healthy control group and are often associated with epilepsy and behavioral problems.

1. Introduction

Several studies in typically developing children have reported that both the type and the prevalence of sleep disorders can vary according to age [1–3]. Infants and toddlers often have difficulties settling to sleep or sleeping through the night. Pre-adolescents show an increased incidence of disorders such as sleep-walking, nightmares, bruxism, and enuresis whereas adolescents are mainly affected by insomnia and daytime sleepiness [1]. A recent review also highlights differences in sleep patterns from the first months of life to adolescence in terms of sleep duration, night waking, sleep latency, longest sleep period, and number of daytime naps [2]. Other authors also report a higher prevalence of dyssomnias and parasomnias in preschool children [3] with a possible correlation with childhood behavior problems, mental retardation or other medical problems [1].

In children with CP, sleep disturbances are more frequent than in typically developing children [4–10], possibly related to motor impairment, pain, behavioral problems or epilepsy. Few studies are available on sleep disturbances in children with CP at different ages. These studies mainly assessed children over 6 years of age and only one included pre-school CP children using questionnaires [10].

The aims of the present study were: (1) to analyze the prevalence of sleep disorders in pre-school children with CP using a structured questionnaire validated for this age; (2) to evaluate differences with typically developing children matched for age and gender; (3) to analyze the relations between sleep disorders and motor, cognitive and behavioral problems.

2. Methods

The children included in this study are part of a collaborative prospective project on families of children with CP regularly followed at the Child Neurology Unit of the Catholic University of Rome and

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at the Neurological Institute Besta in Milan, Italy, between January 2010 and December 2012. The study protocol was approved by the Ethics Committee of the Institutions and informed consent was obtained from parents.

CP was defined as a group of disorders of the development of movement and posture attributed to non-progressive disturbances that occurred in the developing fetal or infant brain, causing activity limitation [11]. Clinical diagnosis was based on the predominant type of motor impairment and classified according to the criteria proposed by Himmelmann et al. [12]. The children were divided into four groups according to the type of CP: diplegia, hemiplegia, quadriplegia, and dyskinesia.

In order to have a homogeneous cohort, we only included children with no parental history of a severe or chronic medical condition (e.g. stroke, diabetes) or psychological disorder. The inclusion criteria were: a diagnosis of CP, age between 3 and 6 years, and a detailed cognitive and motor assessment. A recent study in older children has been published by our group using the same methodological approach [7].

Sleep disturbances were assessed using the Sleep Disturbance Scale for Children (SDSC) validated for pre-school children [13]. It investigates the occurrence of sleep disorders during the previous 6 months, and contains 26 items in a Likert-type scale with values 1–5 (higher numerical values reflect a higher clinical severity of symptoms). The sum of scores provided a total sleep score with a possible range from 26 to 130; a T-score of more than 70 (>95th centile) was regarded as abnormal, and a score of 70 or less as normal.

The factor analysis yielded six sleep disturbance factors representing the most common areas of sleep disorders in pre-school children: parasomnias (PAR) related to arousal disorders (sleepwalking, sleep terrors), nightmares, and sleep–wake transition disorders; difficulty in initiating and maintaining sleep factor (DIMS) related to sleep duration and latency, problems in falling asleep and night awakenings; sleep disordered breathing (SDB); disorders of excessive somnolence (DOES) related to daytime somnolence and sleep attacks; sleep hyperhydrosis (SHY) referred to falling asleep as well as night sweating and nocturnal hyperkinesia; and non-restorative sleep (NRS) concerning items like “the child is unusually difficult to wake up in the morning” or “the child awakes in the morning feeling tired.”

This questionnaire was distributed to the primary caregiver of the children during the routine neurological assessment in our units. Demographic and health questions are not included in the SDSC and were included as a separate partially structured demographic questionnaire. The requested data were gender, age, family status, school education and health status of parents, and age and gender of children. The demographic data were only used to assess statistical differences between the groups on the requested information.

All the children were also screened for the presence of epilepsy, controlled or intractable/active when the seizures fail to come under control with treatment, and antiepileptic therapy.

The SDSC was further distributed to the primary caregivers of a group of children recruited via nurseries and considered as a control group. Questionnaires were filled out by the mothers during the school hours under the supervision of the researchers that distributed the questionnaires (authors: CB, EM, SS); no missing values were reported. All children in the control group attended regular classes in mainstream nursery school and those with obvious or reported sign of mental, developmental or physical disabilities based on school medical records or receiving on-going prescription medication (antiepileptic drugs, antihistaminic drugs, benzodiazepine, melatonin) were excluded.

All children in this study also underwent a comprehensive assessment of motor, cognitive, and behavioral abilities.

Motor function was evaluated using the Gross Motor Function Classification System (GMFCS) [14] to classify each child’s level of gross motor function with skill levels from I to V, assessing the children’s gross motor function by observation.

Cognitive function was measured using the Wechsler Preschool and Primary Scale of Intelligence, 3rd Edition (WPPSI-III) [15] for children between 3 and 6 years of age. The test was performed by a trained psychologist.

Child behavior was assessed using the Child Behaviour Check List (CBCL) [16,17]. In this test behavior problems are reported by the child’s primary caregiver (the person who is most responsible for the day-to-day decision making and care of the child). The CBCL consists of 118 items on which parents rate their child’s behavior by using 3-point scales: 0 (not true), 1 (somewhat or sometimes true), and 2 (very true or often true). The CBCL provides a total behavior problems score, 2 second-order factor scores (internalizing problems, externalizing problems), and 8 syndrome scores (aggressive behavior, anxious/depressed, attention problems, delinquent behavior, social problems, thought problems, withdrawn, and somatic complaints). Raw scores on each clinical factor were transformed to T-scores based on published norms. Scores >63 are considered abnormal, scores between 60 and 63 as borderline, and scores <60 normal.

2.1. Statistical analysis

Data were presented as mean values (standard deviations [SDs]) for continuous variables normally distributed, median (interquartile range) for non normal continuous data, and count and percentages for categorical variables. The comparisons between the different types of CP for the continuous variables (age of children, cognitive assessments, scores on SDSC total and 6 factors and CBCL scales) were performed using the Kruskal–Wallis equality-of-populations rank test; the comparisons for the categorical variables (gender, epilepsy controlled or active) and GMFCS scores were performed with the Fisher’s exact test.

The comparison between children with CP and control group for SDSC total and the six factor scores and age was performed using the non-parametric Mann–Whitney U test; the comparisons for the gender were performed with the Fisher’s exact test. The association between an abnormal total SDSC score and the physical parameters (sex, age, CP type, developmental delay, GMFCS level, epilepsy, and abnormal CBCL scores) were performed, and reported as crude odds ratios (OR) with their 95% confidence intervals (CI).

Multivariate analysis was conducted by logistic regression to define the role of specific factors that may affect an abnormal total SDSC score. All the significant variables obtained after univariate analysis were entered into the initial model. Results of the logistic regression are expressed as OR with 95% CI.

A two-tailed value of $P < 0.05$ was considered significant. Statistical analysis was performed using the “Stata Statistical Software: Release 10” (StataCorp LP, College Station, Tx).”

3. Results

During the study period, 100 children with CP (52 M, 48 F) and the primary caregiver fulfilled the inclusion criteria: 32 children presented diplegia [14,18], 34 hemiplegia [16,18], 29 quadriplegia [11,19] and 5 dyskinesia [5]. The mean age was 3.8 ± 0.8 years (range 3–5 years). No statistical difference was found among the various types of CP for both mean ages of children (diplegia 3.8 ± 0.8 ys; dyskinesia 4 ± 1 ys; hemiplegia 3.9 ± 0.8 ys; quadriplegia 3.9 ± 0.8 ys) or gender .

Thirty-two children (32%) were affected by epilepsy (18 with quadriplegia, 10 hemiplegia, 4 diplegia) and were all receiving

Table 1
Distribution of children based on motor and cognitive functioning.

	Diplegia	Hemiplegia	Quadriplegia	Dyskinetic	Total
GMFCS level					
I	11	29	0	0	40
II	14	5	2	2	23
III	7	0	7	1	15
IV	0	0	5	1	6
V	0	0	15	1	16
IQ					
Normal	22	22	2	0	46
Borderline	4	0	0	1	5
Mental retardation	6	12	27	4	49

Abbreviations: GMFCS: Gross Motor Function Classification System; IQ: intelligence quotient.

antiepileptic medication; 15 out of 32 children showed intractable/active epilepsy (12 quadriplegia, 1 hemiplegia, 2 diplegia).

The questionnaire was also completed for 100 healthy children (55 males) with a median age of 3.9 ± 0.8 years (range 3-5 years). This control group presented the same distribution of age and gender compared with CP group ($P > 0.05$).

3.1. Motor and cognitive function

Table 1 reports the distribution of motor and cognitive function in children with CP.

In the intergroup comparison of motor functions, there was a statistical difference on distribution of GMFCS level ($P < 0.001$): children with hemiplegia were mainly in level I at GMFCS; children with diplegia were distributed in level I, II, and III, whereas children with quadriplegia were mainly in level IV and V.

In the intergroup comparison, there was a statistical difference on the distribution of cognitive level ($P < 0.01$): children with hemiplegia and diplegia reported a normal IQ in almost the 50%, whereas children with quadriplegia showed a mental retardation in more than 90%.

3.2. CBCL results

Details of the scores of the CBCL, reported by the primary caregiver (mother in all cases) are reported in Table 2. In the intergroup comparison, no statistical differences ($P > 0.05$) were observed on internalizing, externalizing or total CBCL scores. Almost all children with diplegia, hemiplegia and quadriplegia had median total internalizing and externalizing scores falling within the normal range; the whole group of children with CP showed pathological scores in the 14%, 13%, and 15% respectively for total, internalizing, and externalizing scores.

3.3. SDSC results

An abnormal total sleep score (>70) was found in 13 children with CP; 35% had an abnormal score on at least one SDSC factor: PAR 9%, DIMS 18%, SDB 13%, DOES 5%, SHY 9%, and NRS 5%.

In the intergroup comparison between the four types of CP, no significant differences ($P > 0.05$) were reported for SDSC total or factor scores (Table 3).

In the univariate analysis, an abnormal SDSC total score was significantly associated with the presence of quadriplegia, presence of epilepsy (controlled or active), CBCL scores (internalizing and total score), and level V on GMFCS (Table 4).

On multivariate analysis (Table 5), an abnormal total SDSC score was significantly associated with the presence of pathological internalizing scores on CBCL and active epilepsy.

Table 6 reported the incidence of abnormal SDSC scores related to the associated clinical factors (cognitive-motor function, behavior and epilepsy).

3.4. Comparison of sleep disorders between CP and control group

CP group had higher significant median scores on SDSC total (55.6 vs. 37.4, $P < 0.00001$), PAR (54.8 vs. 50.2, $P < 0.01$) and DIMS (57.5 vs. 48.7, $P < 0.00001$), whereas no statistical differences were reported on SDB (51.7 vs. 48.5), DOES (49.7 vs. 49.1), SHY (52.2 vs. 50.0) and NRS (50.9 vs. 50.6,) (Table 3).

4. Discussion

Different studies have considered sleep disorders in children with CP in both school age children and adolescents using structured questionnaires [4–10]; less has been reported for younger children at preschool ages [10].

Table 2
CBCL findings in the different groups of CP.

	Diplegia	Hemiplegia	Quadriplegia	Dyskinetic	*P*
CBCL internalizing	52 (34–75)	49 (34–88)	50 (30–74)	45 (34–40)	>0.05
CBCL externalizing	50 (35–69)	53 (30–90)	46 (30–69)	53 (41–49)	>0.05
CBCL total	53 (36–71)	53 (30–95)	50 (33–71)	53 (45–48)	>0.05

Median (interquartile range).
Abbreviation: CBCL: Child Behaviour Check List.

Table 3
SDSC total and factor scores in children with cerebral palsy.

	Total	PAR	DIMS	SBD	DOES	SHY	NRS
Diplegia	55.4 ± 11.5	54.2 ± 12.7	57.3 ± 13.1	53.1 ± 13.9	49.8 ± 11.9	52.1 ± 11.6	50.3 ± 10.1
Dyskinesia	48.2 ± 6.1	46.6 ± 3.5	50.2 ± 6.1	43.4 ± 3.3	45.0 ± 0.0	54.6 ± 10.0	48.8 ± 7.2
Hemiplegia	53.6 ± 9.6	54.3 ± 11.1	54.8 ± 12.2	49.4 ± 11.4	47.2 ± 6.6	51.4 ± 12.2	50.8 ± 9.1
Quadriplegia	59.3 ± 17.0	57.4 ± 11.3	58.0 ± 17.5	54.4 ± 18.1	53.1 ± 16.2	52.7 ± 13.8	52.1 ± 13.7
CP group	55.6 ± 12.8	54.8 ± 11.6	57.5 ± 14.3	51.7 ± 14.3	49.7 ± 11.8	52.2 ± 12.2	50.9 ± 10.7
Control group	37.4 ± 7.3*	50.2 ± 8.8*	48.7 ± 8.6*	48.5 ± 10.4	49.1 ± 8.3	49.9 ± 9.6	50.6 ± 9.4

Abbreviations: PAR: parasomnias; DIMS: difficulty in initiating and maintaining sleep; SDB: sleep disordered breathing; DOES: disorders of excessive somnolence; SHY: sleep hyperhidrosis; NRS: non-restorative sleep.
* $P < 0.01$ Control group vs. CP group.

Table 4
Univariate analysis of associations between the independent variables and abnormal total SDSC score.

	Crude	*P*
	OR (95% CI)	
Gender		
Male	0.665 (0.170–2.5340)	0.558
Age		
2–3 (baseline)	–	–
4	1.273 (0.323–4.839)	0.766
5	1.074 (0.173–4.758)	1.000
Diagnosis		
Diplegia (baseline)	–	–
Hemiplegia	0.313 (0.032–1.584)	0.208
Quadriplegia	**3.447 (0.874–13.705)**	**0.049**
Dyskinetic	0.000 (0.000–5.234)	1.000
IQ		
Normal (baseline)	–	–
Borderline	0.000 (0.000–4.290)	1.000
Mental retardation	3.258 (0.760–19.455)	0.133
Epilepsy		
Controlled	**4.200 (1.070–17.749)**	**0.023**
Active	**4.813 (1.010–20.544)**	**0.024**
CBCL		
Internalizing	**18.889 (2.215–222.388)**	**0.002**
Externalizing	7.167 (0.084–568.775	0.244
Total	**18.889 (2.215–222.388)**	**0.002**
GMFCS		
I (baseline)	–	–
II	0.246 (0.006–1.868)	0.288
III	1.035 (0.100–5.641)	1.000
IV	1.367 (0.027–13.810)	0.576
V	**6.600 (1.473–27.898)**	**0.005**

Significant results ($P < 0.05$) are indicated in bold type.
Abbreviations: OR: odds ratio; CI: confidence interval; IQ: intelligence quotient; CBCL: Child Behaviour Check List; GMFCS: gross motor function classification system.

Our findings confirm that, at pre-school age, children with CP already present a high incidence of sleep disorders, with over 30% presenting at least one sleep disorder. In our cohort DIMS and SDB were the most frequent abnormalities. There was no significant difference in SDSC total or factor scores among the four types of CP or according to the age (3–5 years). In contrast, the presence of epilepsy appeared to significantly increase the risk of abnormal SDSC total score. We found an abnormal SDSC total score in 33% of children with CP and active epilepsy vs. 17% in children with CP and controlled epilepsy and 7% in children with CP without epilepsy. This is not surprising as seizure can impair the quality and restorative value of sleep, modifying circadian sleep–wake rhythms and total duration of sleep. Other effects on sleep may also be related to antiepileptic medication [19] such as barbiturates (daytime sleepiness), phenytoin (insomnia) or benzodiazepines (oversedation) or to a possible effect of the seizures per se. In agreement with previous studies [1,4,7], these findings in children with CP at older age confirm this association even at pre-school age.

Table 5
Multivariable analysis of variables associated with an abnormal total SDSC score.

	OR (95% CI)	*P*
Quadriplegia	0.935 (0.08–10.380)	0.956
Epilepsy – Active	**14.04 (1.255–157.078)**	**0.032**
Epilepsy – Controlled	0.866 (0.114–6.577)	0.890
CBCL Internalizing	**71.266 (4.137–1227.675)**	**0.003**
CBCL Total	6.236 (0.482–80.536)	0.161
GMFCS Level V	5.647 (0.545–58.433)	0.146

Significant results ($P \leq 0.05$) are indicated in bold.
Abbreviations: OR: odds ratio; CI: confidence interval; IQ: intelligence quotient; CBCL: Child Behaviour Check List; GMFCS: gross motor function classification system.

Table 6
SDSC scores and associated clinical factors.

	SDSC total	SDSC total
	≥70	*<70*
GMFCS		
Level I	3	37
Level II	1	22
Level III	2	13
Level IV	1	5
Level V	6	10
IQ		
Normal	2	38
Borderline	0	5
Mental retardation	11	44
Epilepsy		
Absent	5	62
Controlled	3	15
Active	5	10
CBCL total		
Normal	6	80
Abnormal	7	7

Abbreviations: CBCL: Child Behaviour Check List; GMFCS: gross motor function classification system; IQ: intelligence quotient.

Our data further underline that behavioral problems are also related to sleep disturbances in young children with CP [18,20]. Abnormal SDSC total scores were associated with internalizing disorders such as withdrawn, somatic complains, anxiety/depression that, even in the general population, are known to be associated to sleep problems [21], whereas a poor relation was observed between sleep disorders and externalizing factors (delinquent and aggression behavior).

The main differences between study and control group were related to PAR, DIMS and total scores. PAR and DIMS had frequently abnormal scores in healthy children at 3–5 years [13] but they were even more frequent in the CP group. Other studies confirm that nightmares, sleep–wake transition disorders, problems in falling asleep, and night awakenings occurred more frequently in CP children with behavioral problems and epilepsy [4,7,9].

We have recently reported the results of a study of sleep disturbances in an older population of CP children [7]. The preschool CP children included in the present study had a slightly lower incidence of abnormal total sleep and factor scores than those assessed at an older age. The results however are not fully comparable because of the differences in the factorial structure of the SDSC applied at different ages [13,22]. Nevertheless a comparison between the parts of the questionnaire with similar structure, such as the SBD and SDSC total scores, showed that the SBD had higher scores in the older children. Sleep-related breathing disturbances are common in children with CP because of motor impairment associated with swallowing difficulties and other worsening factors like adenotonsillar hypertrophy, gastro-esophageal reflux [1,23,24], subsequent upper airway obstruction, and mixed sleep apnea. The higher incidence of SBD in older CP children, already reported in the only previous study comparing pre-school and school age CP children [10], is probably related to the unfavorable progress of airway obstruction due to increasing muscular hypotonia with age, as also suggested by the higher incidence of clinical or surgical intervention in children older than 5 years [24]. Finally both older [7] and younger CP children reported similar clinical factors associated with an abnormal SDSC total score as epilepsy and behavioral problems, confirming that they are important risk factors for the development of sleep disorders irrespective of the age.

Our study therefore suggests that sleep disorders are already present in children with CP at pre-school age. The results should however be interpreted with caution as some items, especially those

assessing non-restorative sleep, may not be entirely applicable to children with severe motor disorders.

Even with this limitation, our results suggest that the SDSC provides a useful screening for the evaluation of comorbid sleep disorders in CP, identifying the children who need more detailed assessment. As sleep disturbances have a strong impact on the quality of life of children and their families [6,9], the early identification of sleep disorders could allow an effective age-related treatment. Further studies assessing other clinical aspects like pain, feeding difficulties, gastrostomy tube feedings, suctioning, and repositioning at night and a more structured in-depth interview with objective assessments investigating the child's experienced burden or actigraphy may provide additional information on the possible effects of these variables on sleep and improve treatment strategies.

Conflict of interest

The ICMJE Uniform Disclosure Form for Potential Conflicts of Interest associated with this article can be viewed by clicking on the following link: http://dx.doi.org/10.1016/j.sleep.2014.05.008.

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